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CASE REPORT

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Embolization of a large, symptomatic splenic artery pseudoaneurysm

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Summary

Background: Splenic artery aneurysm is the third most common abdominal aneurysm. Most often it is due to pancreatitis. There were only 19 cases of aneurysms larger than 5 cm in diameter described in the literature. Management of splenic artery aneurysms depends on the size and symptoms. Invasive treatment modalities involve open procedures and interventional radiology methods (endovascular).

Case Reports: A 44-years-old male with chronic pancreatitis, in a gradually worsening general condition due to a large splenic artery aneurysm, was subjected to the procedure. Blood flow through the aneurysm was cut-off by implanting a covered stent between celiac trunk and common hepatic artery. Patient's general condition rapidly improved, allowing discharge home in good state soon after the procedure.

Conclusions: Percutaneous embolization appears to be the best method of treatment of large splenic artery aneurysms. Complications of such treatment are significantly less dangerous than those associated with surgery.

MeSH Keywords: Splenic Infarction • Embolization, Therapeutic • Stents • Aneurysm, False

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Background

Splenic artery aneurysm is the third most common abdominal aneurysm following abdominal aortic aneurysm and iliac artery aneurysm [1]. It is four times more frequent in women, but men are three times more likely to have a rupture [2].

Factors leading to development of splenic aneurysm include: hypertension, portal hypertension, liver cirrhosis, liver transplantation, and pregnancy. Less common causes include: arterial fibrodysplasia, vascular collagen defect, alpha-1 antitrypsin deficiency, inflammation and infections. Atherosclerosis plays a smaller role than in the aneurysms of large vessels [2–4]. Splenic artery aneurysms reach a size of 2–9 cm, but usually do not exceed 3 cm. There may be multiple aneurysms. They are most often located in the distal part of splenic artery [5]. About 2–3% of splenic artery aneurysms rupture. The risk of rupture increases in the presence of portal hypertension, following liver transplantation and during pregnancy [6].

Some asymptomatic splenic artery aneurysms are diagnosed incidentally during ultrasound examination of other organs. In case of suspicion of splenic artery aneurysm the diagnostics should be broadened to include US Doppler, angio-MRI and angio-CT examinations. Subtraction angiography is the gold standard and may be performed together with a therapeutic procedure [7–9].

Management of splenic artery aneurysms depends on their size and symptoms. True asymptomatic splenic artery aneurysms up to 2 cm in diameter should be followed up. Invasive treatment is indicated in asymptomatic aneurysms larger than 2 cm, symptomatic aneurysms, pregnant women, women of childbearing age, patients referred for liver transplantation and patients with portal hypertension [2].

Figure 1. CT before the stent – graft implantation procedure, with contrast medium, arterial phase. Contrast medium and parietal thrombus filling partially lumen of aneurysm.

Treatment of splenic artery aneurysms, both true aneurysms and pseudoaneurysms, may be performed using two methods: open surgery or endovascular procedure.

Case Report

A 44-years-old male with chronic pancreatitis complicated by pancreatic cysts, with history of acute pancreatitis, was admitted to an Internal Medicine Department with symptoms of upper gastrointestinal tract bleeding. Patient suffered from diabetes, malnutrition and passed tarry, bloody stool. Gastroscopy did not reveal the source of bleeding. However, it did reveal esophageal varices. A colonoscopy also did not show a potential source of bleeding. Patient reported epigastric pain radiating to the interscapular region. Physical examination revealed a pulsating pathological mass in the entire epigastrum. The diagnostics was broadened to include imaging studies. Ultrasound examination showed a cystic lesion, 93×74 mm, with blood flow and perimural thrombus visible inside – such picture might correspond to splenic artery aneurysm. Computed tomography with contrast confirmed the diagnosis of splenic artery aneurysm, 128×89×100 mm in diameter (Figure 1). The aneurysm displaced an atrophic pancreas with calcifications and a cyst in its tail, and compressed the stomach. Dilated portal vessels exhibiting signs of portal hypertension and celiac trunk were also displaced.

Two months before admission patient was hospitalized in the General Surgery Department due to abdominal trauma. Splenic artery rupture was suspected based on angio-CT picture and patient underwent surgery. No signs of aneurysm rupture as well as intra- or extraperitoneal bleeding were noted during the laparotomy. Mesentery and epigastrum were enfolded by fibrous adhesions, which in the opinion of the operating surgeon would have prevented the aneurysm from rupturing into the peritoneal space. On this basis and due to the difficulty of the operation and high periprocedural risk no attempt at removing the aneurysm was made at that time.

Patient's general state gradually deteriorated despite intensive conservative treatment: significant hypovolemia, hypotonia, anemia that could not be corrected with blood

Figures 2. (**A, B**) Digital subtraction angiography of celiac trunk. Rapid contrast inflow to aneurysm.

transfusions, labile glycaemia (blood glucose levels reached 400 mg%). Patient developed a hyponatremia with neurological manifestations. Moreover, metabolic acidosis was noted. Patient was referred to the Vascular Surgery Department for further treatment. Due to severe general state and lack of signs of aneurysm rupture patient was qualified for endovascular treatment. Celiac trunk angiography was performed by puncturing the femoral vein using Seldinger method and splenic artery aneurysm, 75×47 mm in size, was visualized (Figure 2A, 2B). Attempts at cannulation of the splenic artery were unsuccessful due to atypical branching (under very sharp angle) of splenic artery from celiac trunk. Therefore, the stentgraft (FLUENCY plus Vascular Stent Graft, 7 mm in diameter, 60 mm in length) was implanted in the celiac trunk with its distal end located in the common hepatic artery. Inflow into the splenic artery was practically cut off and only minute blood flow into the aneurysm was demonstrated (Figure 3).

Gastrointestinal bleeding subsided. Abdominal pain was significantly reduced and patient's general condition improved rapidly. Electrolyte and metabolic imbalances as well as anemia were corrected. Patient regained appetite. Physical examination revealed involution of abdominal mass. No pulsation was noted. A control US examination demonstrated a reduction of aneurysm size to 80×60 mm and lack of blood flow within its lumen. On the 6th day after surgery patient presented with fever of up to 40°C – postembolization syndrome was diagnosed.

Computed tomography was performed 12 days after stentgraft implantation (Figures 4 and 5). Partially preserved blood flow through splenic artery was demonstrated. However, no inflow of contrasted blood was visualized

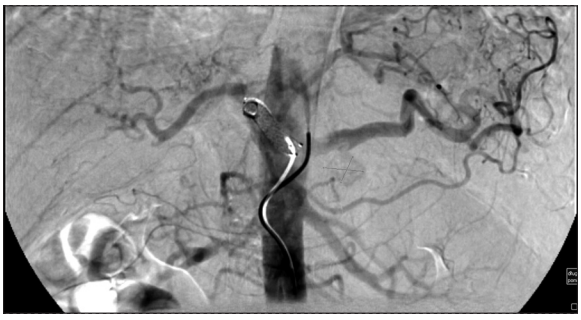

Figure 3. Control digital subtraction angiography immediately after the implantation of the stent – graft. Trace contrast inflow to aneurysm, splenic artery blood flow partially preserved.

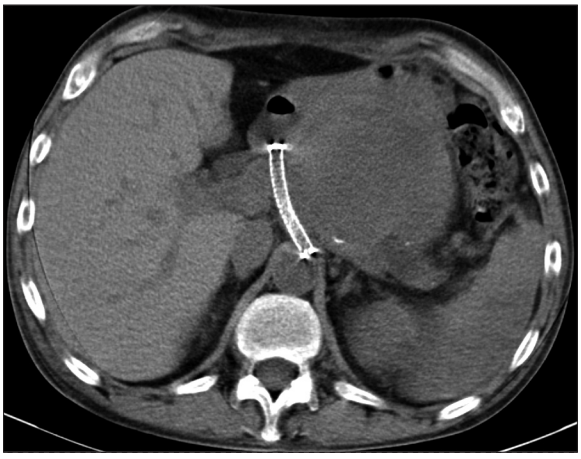

Figure 4. Control CT 12 days after procedure, without contrast. Well visualized the stent – graft in celiac trunk – common hepatic artery.

within the lumen of the aneurysm (Figure 5) and its dimensions decreased to 109×80 mm. The spleen became enhanced in only 50% following administration of contrast medium – partial splenic infarction was diagnosed (Figure 6).

Patient's general condition improved significantly. Fever subsided. High-protein diet was commenced. Patient was discharged home in good general state with recommendations to perform follow-up angio-CT examinations according to Leffroy's scheme: at 6 months, 12 months and once a year [10].

Discussion

Splenic artery pseudoaneurysms with walls consisting of two layers only (intima and media) are not as common as true aneurysms. Literature in English describes only 200 such cases [11]. The most common cause of their development is chronic pancreatitis or, somewhat less often, acute pancreatitis. Pancreatic enzymes in acute pancreatitis cause necrotizing vasculitis, changing wall architecture and leading to defragmentation of collagen fibers [12]. In case of pancreatic pseudocysts pancreatic enzymes are released beyond the cyst and damage vascular wall, while blood gathers within the cyst lumen. According to another theory, direct pressure exerted by the cyst on a vascular wall may cause necrosis and lead to pseudoaneurysm

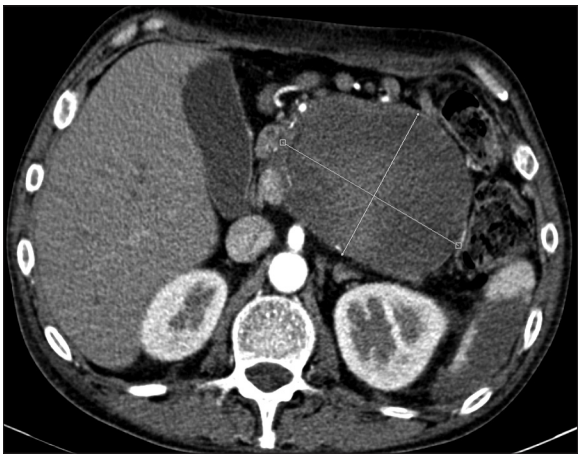

Figure 5. Control CT 12 days after the procedure, with contrast, arterial phase. No inflow of contrast to aneurysm.

Figure 6. Control CT 12 days after the procedure, with contrast, arterial phase. Splenic infarction.

formation [13]. Pseudoaneurysms may also form as a result of iatrogenic injury to splenic artery or, rarely, due to gastric ulcer [11].

Size of splenic artery pseudoaneurysms may very – from 0.3 to 17 cm. There were only 19 described cases of aneurysms larger than 5 cm in diameter and they were called giant pseudoaneurysms. In 41% they coexist with postinflammatory cysts in the course of chronic pancreatitis. Splenic artery pseudoaneurysms are almost always symptomatic. The most common symptoms include: pain – 29.5%, bloody and tarry stool – 26.2%, hemorrhage into pancreatic duct – 20.3%, vomiting blood – 14.8%. Hemorrhage into pancreatic duct, peritoneum, retroperitoneal space or neighboring organs – stomach or transverse colon - is the main complication of pseudoaneurysms [11]. The risk of splenic artery pseudoaneurysm rupture is as high as 37%. Mortality in an untreated rupture reaches 90%. Both small and large pseudoaneurysms rupture [14]. Splenic recess is a particularly dangerous location for such lesions. Pseudoaneurysms located there may rupture during an embolization attempt [15].

Encouraging results of endovascular treatment and a small proportion of complications indicate endovascular

procedures as first-line treatment for splenic artery aneurysms. In case of large pseudoaneurysms this method may not suffice [16]. Masatsugu from Japan presented 3 cases of bleeding pseudoaneurysms following acute pancreatitis, each after unsuccessful attempts at endovascular treatment. Recurrent hemorrhage occurred after successful embolectomy in one case. Authors suggest that in case of recurrent bleeding or after an unsuccessful intervention immediate, aggressive surgery with resection of the spleen and pancreatic tail is indicated [17]. Due to frequent recurrent bleedings following pseudoaneurysm embolization (66.7%) Hsu et al. recommend immediate surgery following endovascular treatment in each case of bleeding pseudoaneurysm [18].

Partial splenic infarction occurred in the case of presented patient, but hemorrhage into the gastrointestinal tract was effectively managed. No recurrent bleeding was noted during a 2-month follow-up period. Classical surgery would have been burdened with extremely high complication risk due to previous operations and portal hypertension.

Classical surgical approach depends on the type of aneurysm, its size and location. When the aneurysm is located in the proximal and middle 1/3 of splenic artery, it should be removed, the artery ligated and spleen should be spared. When the aneurysm involves distal 1/3 of splenic artery splenectomy should be performed together with aneurysm resection. In cases of symptomatic true aneurysms and pseudoaneurysms surgery should be extended to include splenectomy and partial pancreatectomy [19].

Open surgery carries a high risk of complications (9%) and is associated with perioperative mortality of 1.3% [20].

Endovascular methods of management of aneurysms reduced periprocedural mortality [21,22]. An aneurysm may be treated endovascularly by embolization (sac packing) or by closing its supplying vessel (sandwich occlusion), implantation of an excluding stentgraft into the vessel lumen, while maintaining its patency [10,23]. Embolization of aneurysmal sac is performed using coils, detachable balloons, gel foam or hemostatic sponges [20]. Vessel that supplies the aneurysm may be occluded with coils, cyanoacrylate glue or through stentgraft exclusion– as in the case of described patient [20,23,24].

The number of complications decreased significantly in the course of endovascular treatment. However, less severe complications such as splenic infarction or postembolization syndrome presenting with pain or elevated temperature, are quite often observed [10,24,25].

Xin described results of endovascular management of 12 true aneurysms and pseudoaneurysms, with a 100% efficacy. Splenic infarction and postembolization syndrome were observed in 66% of cases [24].

Loffroy presented endovascular treatment of 17 splenic artery aneurysms. Efficacy reached 94%. Splenic infarction was noted in 25% of patients [10].

On the other hand, Yamamoto was successful in 88% of embolizations of 16 aneurysms (14 true aneurysms and 2 pseudoaneurysms) [25].

Conclusions

Endovascular procedures are effective in management of splenic artery aneurysms and much safer than open surgery. Technical problems ensuing from atypical anatomical conditions may arise during endovascular procedures, although such situations are rare. In case of giant aneurysms stentgraft embolization seems to be the best treatment method due to a low risk of dangerous complications and high efficacy.

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